

Pure Sylvian Fissure Arteriovenous Malformation

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Pure sylvian fissure arteriovenous malformations(AVMs) are vascular malformations confined to the sylvian fissure without parenchymal involvement. Surgical removal is regarded as difficult because the nidus is located just lateral to important structures such as the basal ganglia and the internal capsule. Because most feeding arteries to the nidus are branches of the middle cerebral artery(MCA), differentiation between these feeders from en passant and normal vessels is of great importance in order to reduce morbidity and mortality from surgical intervention. We report a case of pure sylvian fissure AVM who presented with an intra-temporal lobe hematoma that was located around venous aneurysms distant from the nidus. The clinical characteristics of this AVM and the surgical methods employed to avoid complications are discussed.

KEY WORDS : Sylvian fissure · Arteriovenous malformation · Venous aneurysm.

Introduction

Sylvian fissure arteriovenous malformations(AVMs) are defined as AVMs located in and adjacent to the sylvian fissure^{10,13}. AVMs located entirely within the sylvian fissure without parenchymal involvement are classified as pure sylvian fissure AVMs, originally described by Sugita et al¹⁰. Because pure sylvian fissure AVMs are usually fed by branches of the middle cerebral artery(MCA), it is very difficult to distinguish between feeders from en passant and other normal vessels. Endovascular management is difficult because feeders are usually small in diameter and directly branch off the MCA¹⁰⁻¹³. The nidus is surrounded by important structures such as the basal ganglia and the internal capsule. Inadvertent injury to these structures during the surgery may cause adverse neurological sequelae; therefore, surgical resection is regarded as difficult. Here, we present a case of a pure sylvian fissure AVM whose nidus was completely removed by direct microsurgery at our institution. Additionally, we describe the un-

ique clinical and angiographical characteristics of this AVM and discuss the surgical methods we used to avoid complications.

Case Report

A 33-year old, right-handed man was referred due to severe headaches intractable to analgesic therapy. Computed tomography(CT) scanning revealed a round high-density area in the right temporal lobe (Fig. 1A). Conventional angiography showed a conglomerated vascular tangle in the distal sylvian fissure (Fig. 2A, B, C, D). The nidus was fed by numerous small arteries that were branches of the MCA. A carotid



Fig. 1. A : Computed tomography showing a hematoma in the right temporal lobe. B and C : T-2 weighted axial magnetic resonance images revealing a nidus located at the deep portion in the sylvian fissure (arrow) and a hematoma (arrowhead) located in the temporal lobe distant from the nidus.

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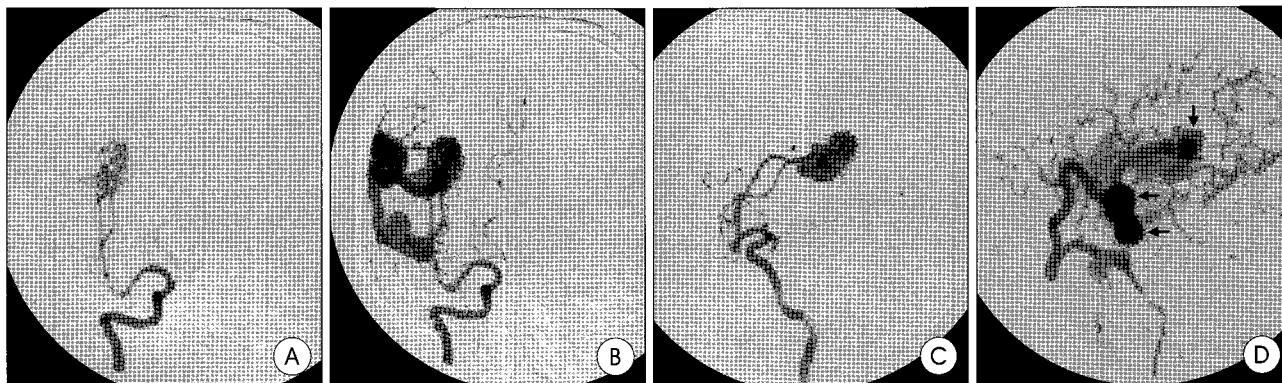


Fig. 2. Early-phase right carotid angiograms with anteroposterior (A, B) and lateral (C, D) views revealing an arteriovenous malformation supplied by the right middle cerebral artery and drained mainly by the superficial sylvian vein which harbors three venous aneurysms (arrows).

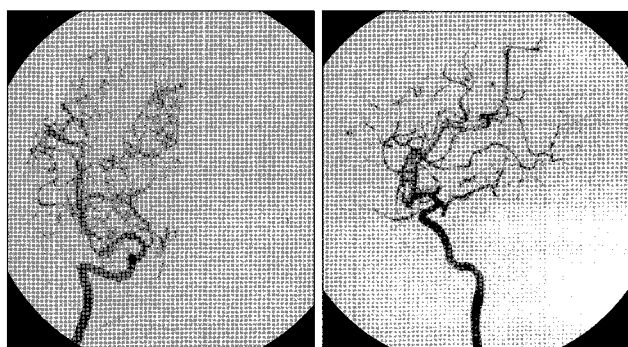


Fig. 3. Postoperative carotid angiograms confirming complete removal of the arteriovenous malformation.

angiogram demonstrated that the feeders were branches of the insular and the opercular segments of the MCA (Fig. 2A, B, C, D). Venous drainage was found to be solely superficial. The sylvian vein was a main draining vein that had three venous aneurysms (Fig. 2B, D). Among these aneurysms, one was located lateral to the nidus and the others were distant from the nidus. Magnetic resonance imaging (MRI) revealed that the hematoma was located around the venous aneurysms distant from the nidus (Fig. 1B, C). Seven days following presentation, the patient underwent craniotomy and removal of the AVM and the hematoma. An extended pterional craniotomy with posterior extension was performed. The sylvian vein was found to be markedly dilated and pulsated with a bright pink color suggesting arterialization (Fig. 4A). The sylvian fissure was opened widely along its entire length (Fig. 4A). Multiple venous aneurysmal dilatations were found. The sylvian vein was then completely dissected in order to allow it to be mobilized freely during surgery. The nidus was found deep in the distal sylvian fissure (Fig. 4B). The AVM was resected by means of a proximal-to-distal and distal-to-proximal direction along the sylvian fissure, not by circumferential direction. The flow resistance and resistive index of the vessels around the nidus were measured with doppler ultrasonography. The nidus and the vessels connecting to the sylvian vein from

the nidus were removed; however, the sylvian vein was preserved. Complete removal of the AVM was thus achieved and confirmed by postoperative angiography (Fig. 3A, B). The patient's post-operative course was uneventful and he was discharged without neurological defects.

Discussion

Pure sylvian fissure AVMs are difficult to manage^{10,11,13}. Furthermore, in the past it was felt that surgical removal of unruptured sylvian fissure AVMs was not justified because of the expected high incidence of morbidity¹³. However, successful removals of AVMs have been reported since the advent of microsurgical instruments and techniques^{2,3,5,10,11,13}. Recently, we have experienced a case of a pure sylvian fissure AVM who presented with an intra-temporal hematoma. Usually, in patients with AVMs, hemorrhage occurs around the nidus or through an associated aneurysm in the feeders^{3,6,7,9}. Venous stenosis is regarded as the key factor leading to hemorrhage^{3,6,7,9}. In our patient, however, hemorrhage occurred around the venous aneurysms (ectasia, dilatation, varix) which were distantly located from the nidus (Fig. 1A, B, C). During surgery, we carefully examined the aneurysms after removal of the hematoma, and were unable to find a ruptured site on the vessel. Pritz⁹ suggested that the presence of a venous aneurysm on a draining vein might increase the risk of AVM rupture. However, the author did not note possible mechanisms why the aneurysm was associated with bleeding. It is well known that the wall of large draining veins of AVMs is much thicker than the wall of normal cortical veins⁵. However, the wall thickness of the venous aneurysm has been reported to be extremely irregular without internal elastic lamina in patient with dural arteriovenous fistula presenting with an intracerebral hematoma⁴. Hamada et al⁴ suggested that abnormally high intracranial pressure might induce chronic endothelial alterations that induce increased endothelial permeability with local destruction of the internal elastic

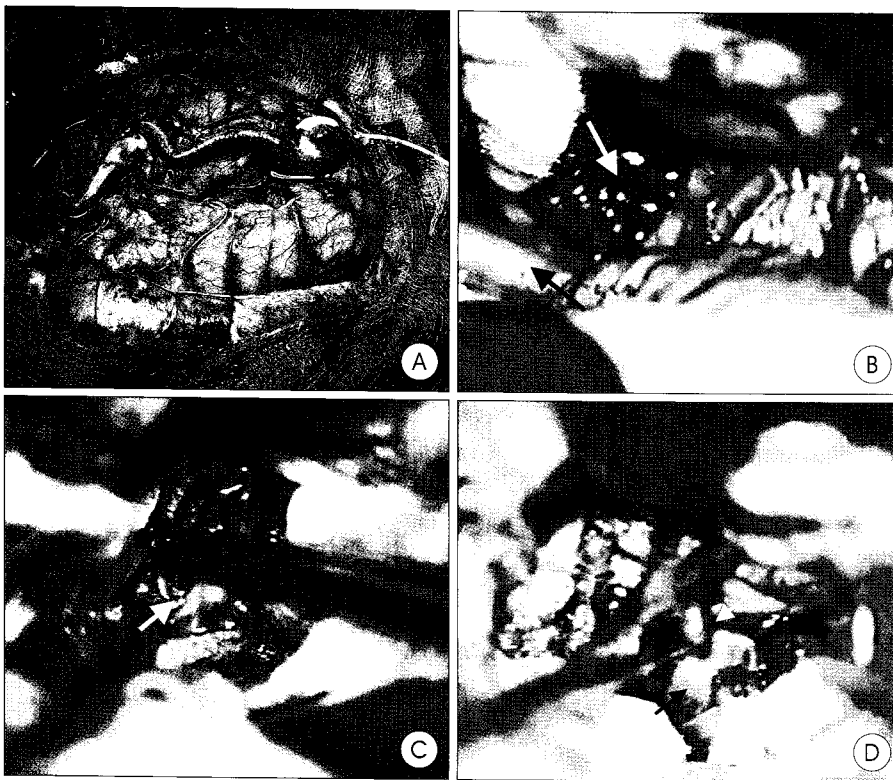


Fig. 4. Operative photographs obtained from the video. A : Photograph showing the surface of the brain. The sylvian fissure was opened widely and the sylvian vein was dissected from the surrounding arachnoid membrane along its entire length. B : Photograph showing a nidus (white arrow) and the superior trunk of the middle cerebral artery (black arrow). C : A feeder branched from the normal artery (arrow) is coagulated by a right-angled bipolar electrocautery. D : Photograph demonstrating the opercular segment of the sylvian vein with a stump of severed feeder (white arrow) and behind it, a large draining vein (black arrow).

lamina. We hypothesize that such an abnormal anatomy of the venous aneurysm may cause a functional disruption of the blood vessel and eventually cause a hemorrhage. In our patient, three venous aneurysms were located at the turning points of blood flow (Fig. 2A, B, C, D). Hemodynamically, with such anatomical features, the large amount of blood from the nidus may have led to the development of the venous aneurysms. Abnormal hemodynamic loading may cause a transient functional disruption of the vessel at a weak point, such as a venous aneurysm, and eventually cause a hemorrhage. Based on surgical findings and angioarchitectural features, we hypothesize that the hemorrhage in our patient may have been caused by a transient functional disruption of the venous aneurysm, not by rupture.

In pure sylvian fissure AVMs, most of the arterial supply to the AVM is directly branched from the MCA (Fig. 4C, D). Distinguishing between transit arteries and feeders is difficult on imaging studies; therefore, the use of endovascular embolization may be limited⁹⁻¹³. Additionally, small-sized vessels including transit arteries and lenticulostriate vessels are susceptible to radiation injury^{1,7,11,13}. The nidus is also frequently long and narrow which requires overlapping isodose centers,

which may increase the risk of radiation injury^{11,13}. Therefore, surgical removal by experienced surgeons is regarded as the best treatment option for pure sylvian fissure AVMs¹³. The key to removal of pure sylvian AVMs with a low incidence of morbidity is to open the sylvian fissure as widely as possible and to use retraction primarily on the AVM itself^{5,10,13}. A dilated sylvian vein is often located superficially and can hide the nidus. It is important to dissect the sylvian vein from the surrounding arachnoid membrane along its entire course, to ensure that the vein is mobile enough to obtain a good surgical field (Fig. 4A). In our case, after skeletonizing the MCA, we used doppler ultrasonography to distinguish between the normal arteries and the feeders. In a study by Dempsey et al², stating that doppler was a useful tool to identify the normal arteries in AVMs, the resistive index(RI) of the vessels around the nidus were estimated. The study

found that the RI of the normal arteries was higher than that of the AVM feeders. The authors concluded that this phenomenon occurred because the normal arteries had a capillary bed, thus causing higher resistance, while the feeders lacked a capillary system². Additionally, the consistency of the RI was maintained despite the direction of the probe in the patients². However, in our patient, the RI was not consistently maintained as the RI changed according to the direction of the probe; thus, the RI was not helpful to us in identifying the abnormal arteries. To differentiate normal from abnormal arteries, we dissected all the vessels thoroughly. As noted in other studies, normal and abnormal arteries must be examined from the beginning to the end of the sylvian fissure^{5,10,13}. To preserve MCA branches that course through the AVM, dissection should extend well beyond the posterior aspect of the AVM into the distal sylvian fissure. We resected the AVM by means of a proximal-to-distal and distal-to-proximal direction along the sylvian fissure, not by circumferential direction. This method proved to be useful in identifying the entire course of the vessel and more accurately determine which vessel could be severed without complication¹³. This dissection method and wide-view perspective can also reduce the risk

of occluding the normal and/or en passant arteries. We preserved the entire course of the sylvian vein, even the portion in the proximity of the resected nidus (Fig. 4D). This portion of the sylvian vein was found to be obliterated spontaneously by thrombosis on the post-operative followup angiogram (Fig. 3A, B). In pure sylvian fissure AVMs, the sylvian vein is usually a main draining vein, including for normal venous blood. Therefore, we suggest that resection of the sylvian vein segment just adjacent to the nidus may not be necessary after complete removal of the nidus.

Conclusion

We experienced a case of pure sylvian fissure AVM at our institution that presented as a hemorrhage associated with a venous aneurysm. The clinical and radiological characteristics of the AVM, as well as the surgical findings and useful techniques to reduce the incidence of morbidity are described.

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